Endoluminal stent graft repair of aortobronchial fistulas

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Objective: **To describe our experience with endoluminal stent graft repair of aortobronchial fistulas.**
Methods: **We reviewed the records of patients treated with endoluminal stent grafting of aortobronchial fistulas at a private teaching hospital. All patients underwent the following diagnostic studies: computed tomography, angiography, bronchoscopy, and transesophageal echocardiography. With standard endovascular techniques, two different devices were implanted.**
Results: **Between March 1997 and October 2000, we treated four patients with postsurgical fistulas. The patients were diagnosed with hemoptysis between 3 and 23 years after aortic replacement grafting for thoracic aneurysms. Diagnostic studies varied in their ability to find the fistula. Transesophageal echocardiography most reliably demonstrated the fistula in the patients. All were successfully treated by exclusion with endoluminal stent grafting. The patients had no complications and no further episodes of hemoptysis.**
Conclusion: **Endoluminal stent grafting of aortobronchial fistulas is feasible and may become the preferred method of management in patients at high risk. (J Vasc Surg 2002;35:387-91.)**

Aortobronchial fistula (ABF) is a rare, potentially life-threatening complication of thoracic aortic replacement surgery. Often seen with substantial hemoptysis, its presence mandates urgent repair. Conventional open surgical correction involves thoracotomy and carries significant morbidity because of the difficulties of operative dissection associated with reoperative surgery.[1-3] The patients often have other severe comorbidities and may have rather poor health. These physiological stresses can be complicated by active hemorrhage and pulmonary compromise. Furthermore, secondary graft infection, prolonged ventilatory compromise, paralysis, kidney failure, and cardiac events are significant potential complications of traditional open repair.[1-5] Endovascular technology as an alternative therapy for ABF has been previously reported.[6-9] We present the successful treatment of four patients with ABFs by endoluminal stent grafting.

METHODS

The records of all patients treated for ABF with endoluminal stent grafting at our institution were reviewed. All patients gave written consent to endoluminal stent of the thoracic aorta and intraoperative studies to evaluate and exclude the fistula in accordance with techniques approved by our institutional review board. The device used in the first case, before the availability of commercially developed grafts, was a composite of woven polyester graft sutured to two Gianturco Z stents (Cook, Inc, Bloomington, Ind). An Excluder self-expandable thoracic graft (WL Gore, Flagstaff, Ariz) was used in the last three cases as part of a single-center investigative device exemption for treatment of thoracic aortic diseases. Devices were selected from hospital stocks on the basis of measurements derived from preoperative computed tomography (CT) scans and aortograms.

The procedures were performed with the patients under general anesthesia by surgeons in a dedicated vascular operative suite with complete fluoroscopic and angiographic capabilities. All patients were given a single intravenous dose of an antibiotic before surgery. Intravascular access was obtained in both groins through the common femoral artery and through the left brachial artery. The common femoral artery determined to have the least amount of occlusive disease was selected for deployment and exposed. The patients were given 5000 units of intravenous heparin for anticoagulation. Arteriographic 5F pigtail catheters were placed in the common femoral artery and brachial artery. Aortography and intravascular ultrasonography were performed to measure the dimensions of the thoracic aorta and distances from the brachiocephalic and visceral vessels. Transesophageal echocardiography (TEE) was used to delineate the exact position of the fistula. If severe atherosclerotic disease within the external iliac arteries prevented the passage of the delivery sheath into the aorta, then a retroperitoneal incision was made, and a Dacron graft conduit was attached to the right common iliac artery for device delivery. A 24F sheath (Keller Timerman Sheath; Cook, Inc) was inserted through the common femoral artery over a stiff 0.035 Amplatz guide wire (Boston Scientific-Meditech, Watertown, Mass) to the level of the thoracic aorta. The device was inserted into the sheath. Temporary

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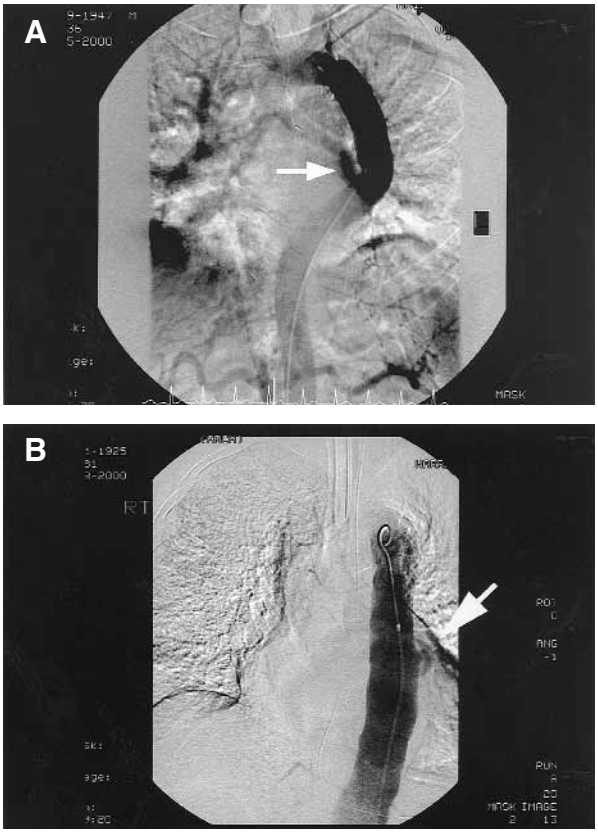

Fig 1. Angiograms show aortobronchial fistulas (**A** and **B**). *Arrows* denote fistulas.

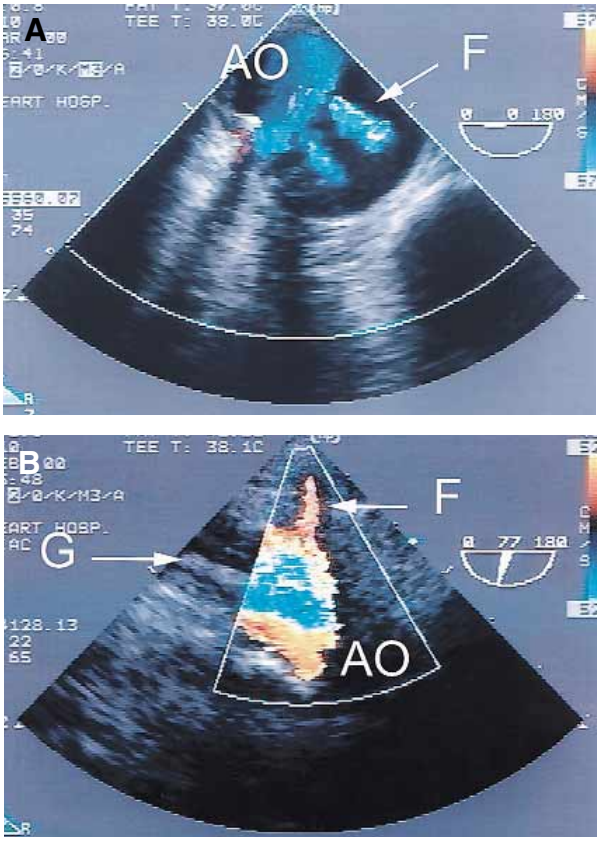

Fig 2. A, Intraoperative TEE of aortobronchial fistula in transverse view. **B,** In longitudinal view, fistula is seen, and stent graft is in position before deployment. *AO,* True aortic lumen; *F,* extraluminal blood flow; *G,* graft; *PSA,* pseudoaneurysm.

hypotension was induced with nitroglycerin. The device was deployed under fluroscopic and TEE guidance at the designated landing zone. Postdeployment balloon dilation was performed only if completion angiography demonstrated an endoleak. Completion arteriography and TEE were performed to assess accurate placement. The patients were kept in the intensive care unit overnight. Postoperative CT scanning of the chest was performed the next day. The patients were discharged from the hospital when they were ambulatory.

RESULTS

Between March 1997 and October 2000, we treated four patients with aortobronchial fistulas by use of endoluminal stent grafts (Table I). All patients had previously undergone thoracic aortic aneurysm repair with graft interposition from 3 to 23 years before onset of hemoptysis. All patients experienced some degree of hemoptysis, ranging from intermittent bloody expectoration to massive hemorrhage, resulting in the need for transfusion and temporary mechanical ventilation. None of the patients was admitted with the sequelae of infection: fever, chills, productive sputum, or leukocytosis. All patients were considered to be "high-risk" surgical candidates because of comorbidities or complications encountered during previous thoracotomies. The patients had hemoptysis for between 10 days to 1 year before undergoing endovascular treatment. In at least two of the cases, the patients had undergone protracted outpatient observations and repeated diagnostic evaluations. In retrospect, these were quite clearly suggestive of the diagnosis but, nonetheless, failed to show an actual fistulous tract. Only when one patient was admitted in respiratory failure with massive active hemorrhage after several prior hospitalizations was the diagnosis entertained. The patient's condition was stabilized, and then she was transferred to our institution for treatment 12 months after her bleeding had begun. Clearly, it may be difficult to diagnose ABF, and a high index of suspicion should be entertained in any patients with a previous thoracotomy and hemoptysis regardless of the lack of evidence with radiographic investigation.

The four most significant diagnostic modalities used for our cases were CT scanning, angiography, bronchoscopy, and TEE (Table II). Although chest radiography and CT scanning were usually the initial radiographic studies, these findings were nonspecific. Pseudoaneurysm, aortic anatomy abnormality, and lung parenchyma com-

Table I. Patient characteristics

Patient	*Age/sex*	*Previous thoracic surgeries*	*Comorbidities*	*Presenting symptoms*	*Interval between last thoracic aneurysm surgery and onset of symptoms*	*Interval between onset of symptoms and treatment*
1	69/F	TAA repair (1974), CABG (1996)	COPD, hypertension, CHF	Hemoptysis	23 years	4 weeks
2	53/M	TAA repair (1984), PSA repair (1989)	Hypertension	Hemoptysis	10 years	7 weeks
3	75/M	Aortic valve replacement (1994), TAA repair (1997)	Hypertension, diabetes, atrial fibrillation, cardiomyopathy	Hemoptysis, Dyspnea, hypoxia	3 years	10 days
4	75/F	TAA repair (1996)	Hypertension, COPD, ulcerative colitis, CHF, steroid dependence, aortic insufficiency	Hemoptysis respiratory failure	3 years	1 year

TAA, Thoracic aortic aneurysm; *CABG,* coronary artery bypass grafting; *COPD,* chronic obstructive pulmonary disease; *CHF,* congestive heart failure; *PSA,* pseudoaneurysm.

Table II. Diagnostic results before treatment

Patient	*Chest CT scan*	*Angiography*	*Bronchoscopy*	*Transesophageal echocardiography*
1	Slight aortic dilation	Fistula visualized; pseudoaneurysm	Blood in left lower lobe	Fistula tract visualized
2	6-cm pseudoaneurysm at the distal graft anastomosis	Fistula suspected	No blood	Fistula tract and pseudoaneurysm visualized
3	6.8-cm pseudoaneurysm at the distal graft anastomosis	No fistula visualized	Blood in left bronchus	Fistula tract and pseudoaneurysm visualized
4	Small saccular pseudoaneurysm at distal graft anastomosis	Fistula visualized; pseudoaneurysm	Blood in both bronchi	Fistula tract and pseudoaneurysm visualized

Table III. Procedural results

Patient	*Type of device*	*Size of device(s)*	*Access*	*Procedure time*	*EBL (mL)*	*Amount of contrast (mL)*	*Follow-up*	*Complications*
1	Gianturco Z stents and polyester graft	34 mm × 10 cm	Femoral artery	140 minutes	200	240	No recurrent hemoptysis, died 2 years after procedure of CHF	None
2	Excluder	28 mm × 7 cm	Femoral artery	160 minutes	300	160	No recurrent hemoptysis at 1 year	None
3	Excluder	34 mm × 20 cm, 40 mm × 10 cm	Femoral artery	120 minutes	100	280	No recurrent hemoptysis at 8 months	None
4	Excluder	34 mm × 20 cm	Iliac artery	140 minutes	400	200	No recurrent hemoptysis at 6 months	None

CHF, Congestive heart failure.

pression were all suggestive findings on each CT scan. Angiography demonstrated a fistulous tract in at least two patients (Fig 1). One patient had an irregular wall in the area of the previous graft suggestive of fistula. Bronchoscopy was performed multiple times in two patients in an effort to find the cause of persistent hemoptysis. The tests showed the presence of blood but failed to reveal the source. Washings and culture specimens were obtained during bronchoscopy for all patients, but none showed any signs of infectious diseases. No patients were previously treated for recurrent pneumonia. Intraoperative TEE yielded the most consistently positive findings, clearly confirming the presence of a fistulous tract because it breached the aortic walls of all four patients (Fig 2,*A*). TEE not only demonstrated the presence of the fistula but also permitted us to guide the device into accurate placement (Fig 2,*B*).

Five devices were successfully deployed in the four patients (Table III). Sizes of the devices were selected according to measurements from CT scanning and intravascular ultrasound scanning. Length of the device was selected so that the proximal and distal graft anastomosis could be excluded. Only one patient was limited by femoral artery size for intravascular access and had to undergo retroperitoneal exposure of the iliac artery with subsequent conduit attachment for device delivery. During deployment, TEE showed the abrupt termination of flow into the pseudoaneurysm in all cases in real time.

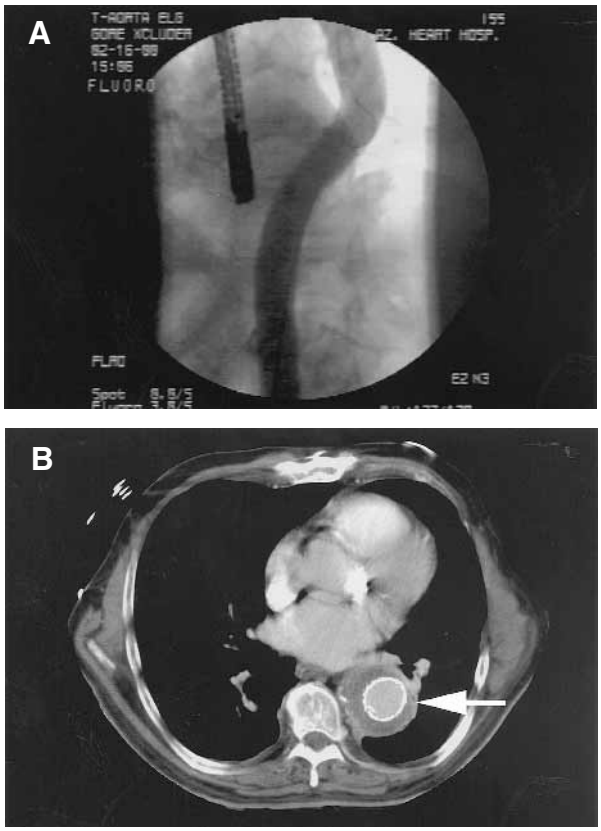

Fig 3. A, Angiogram shows adequate endoluminal stent graft placement. **B,** CT scan of thoracic stent graft after procedure.

Deployment was accurate without device migration. Postdeployment angiography confirmed graft apposition (Fig 3,*A*). Average estimated blood loss was 250 mL, and operative time was 140 minutes.

The patients were extubated within 24 hours and transferred from the intensive care unit the next day. Postoperative CT scanning confirmed accurate placement, and there were no endoleaks (Fig 3,*B*). The patients were usually ambulatory the day after surgery and were discharged by the fourth postoperative day. None had hemoptysis after stent graft placement. No patients had episodes of pneumonia or stent graft infection. There were no operative or postoperative complications. One patient died of chronic congestive heart failure 2 years after undergoing stent grafting. The rest of the patients continue to do well at follow-up after 6 months to 1 year and are seen routinely during clinic visits. All have returned to their normal activities of daily living.

DISCUSSION

Aortobronchial fistula is a rare complication of thoracic aortic surgery. Without prompt surgical treatment, the natural history of the condition is lethal exsanguination. ABF has been reported after the surgical repair of aortic aneurysms, coarctation, patent ductus arteriosus, aortic dissection, valvular heart disease, and other thoracic procedures, with aneurysm repair being the most common culprit.[1-3] The foreign body reaction to prosthetic graft and suture, tracheobronchial compression necrosis, pulsatile pressure erosion, and localized pulmonary infection are all implicated in the pathophysiology of fistula formation.[2] In the presence of an aortic graft, the fistula is associated with a pseudoaneurysm at the anastomotic site—as a result of atherosclerotic disease progression and weakening of the aortic wall. A break in the integrity of the aortic wall results in pseudoaneurysm formation. ABF has been reported from 3 weeks to 23 years after surgery. To date, about 55 cases of postoperative ABF have been reported in the literature. Perioperative mortality rates in patients treated with traditional surgical repair have been reported to be as high as 25% for those with ABF[1,2,5] and 41% in those with pseudoaneurysm.[10] Postoperative morbidity in open surgical repair is substantial and includes prolonged ventilation, pneumonia, multiple organ failure, myocardial infarction, and paraplegia.[4]

The diagnosis of ABF should be given serious consideration in any patient with hemoptysis after a thoracic aortic operation. Such patients warrant immediate diagnostic investigation; however, the actual fistula is often undetected before operation. Chest radiography, CT scanning, angiography, bronchoscopy, chest magnetic resonance imaging, and TEE have all been used to diagnose the lesion, but each has its individual limitations. Chest radiography findings are nonspecific and may demonstrate aneurysm dilation in the presence of pulmonary infiltrates. CT scanning may detect a pseudoaneurysm, periaortic hematoma, and consolidation of adjacent lung. Angiography rarely identifies the fistula. However, it more readily demonstrates the presence of a false aneurysm, ulceration, and aortic irregularities. Bronchoscopy excludes other pulmonary causes of hemoptysis and may show the site of entrance of the fistula into the pulmonary tract. However, bronchoscopy may accidentally dislodge clot within the fistula, resulting in exsanguinating hemorrhage and respiratory arrest.[1,2,5] Magnetic resonance imaging has also been used for diagnosis.[6] In all of our patients, intraoperative TEE demonstrated a breach of blood flow from the aortic graft into a pseudoaneurysm and the presence of the fistulous tract. Although others have expounded the use of CT scanning, angiography, and bronchoscopy in the diagnosis of the fistula, TEE was the most reliable test for our patients.

Once the diagnosis is established, definitive treatment of ABF should be expeditious. Open surgical repair is usually a formidable challenge. The traditional approach involves thoracotomy, aortic cross-clamping, possible cardiopulmonary bypass with concomitant anticoagulation, aorta repair, graft replacement or bypass, and pulmonary tissue dissection in the face of dense adhesions. The patients are often deemed to be at high risk because of their severe comorbidities and the debilitating nature of their condition. The potential for hemodynamic instability in a formal open procedure can be fatal.

Several centers have published their experiences with endoluminal repair of thoracic aortic aneurysms, pseudoaneurysms, dissections, and penetrating ulcers. Preliminary results appear promising. Initial series have reported a 30-day mortality rate of about 10% and an overall technical success rate of more than 90%. The feasibility of this technique continues to be demonstrated in the intermediate results.[11-15] Moreover, experience with endovascular technologies is now yielding a safer procedure. The move to "second-generation" self-expandable stents, familiarity with techniques of accurate device placement, modification in device support systems, and an understanding of the pitfalls of problematic vascular access has diminished the number of perioperative complications.[12] The second-generation devices, such as the Gore Excluder, are easier to deploy, conform more easily to the aortic contour, and are less likely to migrate during placement.

To date, four reports of endovascular repair of ABF in five patients have been published. The cases involved the stent graft closure of ABFs of pseudoaneurysms after aortic coarctation repair, patent ductus arteriosus, aortic surgery for Takayasu's arteritis, and descending thoracic aortic aneurysm replacement. All of these patients had undergone multiple thoracic procedures and were diagnosed with varying degrees of hemoptysis. Each patient was deemed to be at high surgical risk, and therefore endovascular repair was attempted. A self-expanding stent device covered with polyester graft material was deployed in each case with technical success and no further bleeding.

REFERENCES

1. Favre JP, Gournier JP, Adham M, Rosset E, Barral X. Aortobronchial fistula: report of three cases and review of the literature. Surgery 1994;115:264-70.
2. Macintosh EL, Parrott JC, Unruh HW. Fistulas between the aorta and tracheobronchial tree. Ann Thorac Surg 1991;51:515-9.
3. Ishizaki Y, Tada Y, Takagi A, Sato O, Takayama Y, Shimkawa M, et al. Aortobronchial fistula after an aortic operation. Ann Thorac Surg 1990;50:975-7.
4. Ono M, Takamoto S, Kawauchi M, Egami J, Kotsuka Y. Aortobronchial fistula late after transverse arch replacement. Ann Thorac Surg 2000; 70:964-6.
5. Kazerooni EA, Williams DM, Abrams GD, Deeb GM, Weg JG. Aortobronchial fistula 13 years following repair of aortic transection. Chest 1994;106:1590-4.
6. Miyata T, Ohara N, Shigematsu H, Konishi T, Yamaguchi H, Kazama S, et al. Endovascular stent graft repair of aortopulmonary fistula. J Vasc Surg 1999;29:557-60.
7. Campagna AC, Wehner JH, Kirsch CM, Semba CP, Kagawa FT, Jensen WA. Endovascular stenting of an aortopulmonary fistula presenting with hemoptysis: a case report. J Cardiovasc Surg 1996;37: 643-6.
8. Karmy-Jones R, Lee CA, Nicholls SC, Hoffer E. Management of aortobronchial fistula with an aortic stent-graft. Chest 1999;116:255-7.
9. Chuter TA, Ivancev K, Lindblad B, Brunkwall J, Aren C, Risberg B. Endovascular stent-graft exclusion of an aortobronchial fistula. J Vasc Interv Radiol 1996;7:357-9.
10. von Segesser LK, Tkebuchava T, Niederhauser U, Kunzli A, Lachat M, Genomi M, et al. Aortobronchial and aortoesophageal fistulae as risk factors in surgery of descending thoracic aortic aneurysms. Eur J Cardiothorac Surg 1997;12:195-201.
11. Greenberg R, Resch T, Nyman U, Lindh M, Brunkwall J, Brunkwall P, et al. Endovascular repair of descending thoracic aortic aneurysm: an early experience with intermediate-term follow-up. J Vasc Surg 2000;31:147-56.
12. Temudom T, D'Ayala M, Marin ML, Hollier LH, Parsons R, Teodorescu V, et al. Endovascular grafts in the treatment of thoracic aortic aneurysms and pseudoaneurysms. Ann Vasc Surg 2000;14:230-8.
13. Dake MD, Miller DC, Semba CP, Mitchell RS, Walker PJ, Liddell RP. Transluminal placement of endovascular stent-grafts for the treatment of descending thoracic aortic aneurysms. N Engl J Med 1994;331:1729-34.
14. Mitchell RS, Dake MD, Sembra CP, Fogarty TJ, Zarins CK, Liddel RP, et al. Endovascular stent-graft repair of thoracic aortic aneurysms. J Thorac Cardiovasc Surg 1996;111:1054-62.
15. Fann JI, Miller DC. Endovascular treatment of descending thoracic aortic aneurysms and dissections. Surg Clin North Am 1999;79: 551-74.

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